

Review Article

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Interventions to treat fear of childbirth in pregnancy: a systematic review and meta-analysis

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Abstract

Background. Between 5% and 14% of women suffer from fear of childbirth (FOC) which is associated with difficulties during birth and in postnatal psychological adjustment. Therefore, effective interventions are needed to improve outcomes for women. A systematic review and meta-analysis was used to identify effective interventions for treating women with FOC.

Methods. Literature searches were undertaken on online databases. Hand searches of reference lists were also carried out. Studies were included in the review if they recruited women with FOC and aimed to reduce FOC and/or improve birth outcomes. Data were synthesised qualitatively and quantitatively using meta-analysis. The literature searches provided a total of 4474 citations.

Results. After removing duplicates and screening through abstracts, titles and full texts, 66 papers from 48 studies were identified for inclusion in the review. Methodological quality was mixed with 30 out of 48 studies having a medium risk of bias. Interventions were categorised into six broad groups: cognitive behavioural therapy, other talking therapies, antenatal education, enhanced midwifery care, alternative interventions and interventions during labour. Results from the meta-analysis showed that most interventions reduced FOC, regardless of the approach (mean effect size = -1.27 ; $z = -4.53$, $p < 0.0001$) and that other talking therapies may reduce caesarean section rates (OR 0.48, 95% CI 0.48–0.90).

Conclusions. Poor methodological quality of studies limits conclusions that can be drawn; however, evidence suggests that most interventions investigated reduce FOC. Future high-quality randomised controlled trials are needed so that clear conclusions can be made.

Introduction

Research suggests approximately 14% of women are affected by extreme fear of childbirth (FOC), also known as tokophobia (Nilsson et al., 2018; O'Connell, Leahy-Warren, Khashan, Kenny, & O'Neill, 2017). The causes of FOC are thought to be linked to obstetric (Fairbrother, Thordarson, & Stoll, 2018; Haines, Pallant, Karlström, & Hildingsson, 2011; Sydsjö et al., 2013; Sydsjö et al., 2014), psychological (Dencker et al., 2019; Hall, Stoll, Hutton, & Brown, 2012; Jokić-Begić, Žigić, & Nakić Radoš, 2014; Lukasse, Vangen, Åian, & Schei, 2011) and socio-demographic factors (Haines et al., 2011; O'Connell, Leahy-Warren, Kenny, O'Neill, & Khashan, 2019; Ryding et al., 2015). FOC can impact on women's birth choices and outcomes. For example, those with FOC are more likely to opt for an elective caesarean section (CS) (Eide, Morken, & Bærøe, 2019; Ryding et al., 2015; Sydsjö, Sydsjö, Gunnervik, Bladh, & Josefsson, 2012), choose an epidural, have longer labours (Dencker et al., 2019; Logtenberg et al., 2018; Reck et al., 2013) or have an emergency CS (Sydsjö et al., 2012).

FOC has also been found to be related to postnatal psychological adjustment. For example, FOC is associated with postnatal post-traumatic stress disorder (Capik & Durmaz, 2018; Söderquist, Wijma, & Wijma, 2004; Wijma, Ryding, & Wijma, 2002; Wijma, Söderquist, & Wijma, 1997), depression and anxiety (Räisänen et al., 2013; Rouhe, Salmela-Aro, Gissler, Halmesmäki, & Saisto, 2011) as well as antenatal depression and anxiety (Andersson, Sundström-Poromaa, Wulff, Åström, & Bixo, 2004). The long-term impact of FOC on mother–infant outcomes has yet to be investigated; however, research has evaluated the impact of the psychological conditions associated with FOC such as anxiety and depression (Stein et al., 2014). Antenatal mood disorders have been associated with altered patterns of foetal behaviour and heart rate responses (Kinsella & Monk, 2009), more fearful or anxious behaviour in the infant, and increased risk of poor development and adverse child outcomes (Talge,

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Neal, & Glover, 2007). Furthermore, antenatal depressive symptoms have been linked to poorer maternal responsiveness 12 months postnatally (Pearson et al., 2012).

The development of FOC has still yet to be established in the literature; however, a systematic review of 89 studies found that a variety of factors may contribute to the development (Rondung, Thomtén, & Sundin, 2016). In a theory of fear acquisition, Rachman (1977) proposed that fears are developed through three pathways (conditioning, vicarious experiences and transmission of information), and the literature appears to support this model (Rondung et al., 2016). For example, in terms of conditioning, research suggests that negative birth experiences cause future FOC (Lukasse et al., 2011; Lukasse, Schei, & Ryding, 2014; Nilsson, Lundgren, Karlström, & Hildingsson, 2012). Research also suggests that vicarious experiences such as viewing a live birth is associated with a reduction in fear (Stoll & Hall, 2013) and research also suggests that the transmission of information such as negative childbirth stories (Melender, 2002; Tsui et al., 2007) and public discourses of birth (Fenwick, Hauck, Downie, & Butt, 2005; Melender, 2002) also contribute to the development of FOC.

The systematic review also suggested that cognitive aspects may play an important role in FOC (Rondung et al., 2016). For example, women with childbirth fear more commonly report having childbirth-related thoughts compared with women with no fear (Hildingsson, Thomas, Karlström, Olofsson, & Nystedt, 2010). Furthermore, FOC is negatively correlated with childbirth self-efficacy (Beebe, Lee, Carrieri-Kohlman, & Humphreys, 2007) and a woman's appraisal of her ability to cope with stressful situations (Ryding, Wijma, Wijma, & Rydhström, 1998; Söderquist et al., 2004). The review found that behavioural aspects are also important (Rondung et al., 2016) with FOC being associated with avoidance of pregnancy (Tsui et al., 2007), or avoidance of vaginal delivery (Dehghani, Sharpe, & Khatibi, 2014; D'Cruz & Lee, 2014; Matinnia et al., 2015; Nieminen, Stephansson, & Ryding, 2009). Physiological aspects may also be associated with FOC, such as sleep disturbances, tachycardia, tenseness, restlessness and nervousness (Rondung et al., 2016). This complex conceptualisation of the potential causes of FOC suggests that interventions for FOC need to focus on strategies to improve psychological, cognitive, behavioural and physiological aspects of the fear.

To prevent poorer outcomes related to FOC, early detection and evidence-based interventions are key, and should therefore be available to women. Reviews of the evidence suggest that interventions with an educational component may reduce FOC (Moghaddam Hosseini, Nazarzadeh, & Jahanfar, 2018; Stoll, Swift, Fairbrother, Nethery, & Janssen, 2018; Striebich, Mattern, & Ayerle, 2018). However, previous reviews used narrow inclusion criteria such as requiring a minimum score on a FOC measure (Striebich et al., 2018); requiring FOC to be measured twice (Stoll et al., 2018); or only including clinical trials (Moghaddam Hosseini et al., 2018). This means the reviews include very few studies (range = 7–15) and may exclude studies that could provide further insight into effective FOC treatments. Further, the previous reviews did not evaluate evidence quantitatively using meta-analysis. This may have led to a bias in conclusions where interventions that have been more widely examined are more likely to be evaluated as promising. Therefore, this study aimed to conduct a systematic review and meta-analysis to identify the effectiveness of FOC interventions for reducing FOC or rates of caesarean by request during the perinatal period.

Methods

Search strategies

Literature searches and study selection were conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines (Moher et al., 2015) (online Supplementary Table S1). The protocol was registered with PROSPERO (CRD42018093095). Online databases were used to identify papers. Searches were carried out in Cochrane Library (1996–present), EMBASE (1947–present), Healthcare Management Information Consortium (HMIC; 1983–present), Medline (1946–present); PsychInfo (1967–present), PsychARTICLES (1806–present), PubMed (1996–present), SCOPUS (2004–present) and Web of Science (1997–present). Pre-planned searches were carried out using search terms combined with Boolean operators 'OR' and 'AND' (e.g. pregnancy OR perinatal OR postnat* AND fear of childbirth OR tokophobia AND Caesarean OR abdom* deliver* AND counselling OR intervention). Full search syntax can be found in the supporting information. Searches were conducted up to and including June 2019. Two searches were carried out: (1) desire for an elective CS and interventions; (2) FOC and interventions (see Appendix S1). Desire for an elective CS was used as a proxy measure for FOC. Reference lists of all identified papers were also searched.

Inclusion and exclusion criteria

The following inclusion criteria were applied: Participants – women in the perinatal period (the UK definition of perinatal period was used: pregnancy – 1 year after birth; NHS England, 2019) with FOC or tokophobia; Intervention – any intervention that was for women with FOC; Outcome – a measure of FOC or birth outcomes. Studies were excluded if they were conference abstracts, theses, non-English publications or non-empirical papers.

Study selection and data extraction

The results from both searches were imported into Eppi-Reviewer 4. All duplicate papers were removed and studies screened for eligibility based on their title and abstract by one reviewer (RW). A proportion (10% $n = 284$) were double screened by BRG. Reviewers agreed on inclusion/exclusion of studies 99% ($n = 282/284$) of the time. Studies that were not eligible were excluded. Full texts of studies that appeared to meet criteria or where it was unclear were then reviewed by one reviewer (RW) to determine whether they should be included. A proportion (10%, $n = 9$) were double screened by BRG. Reviewers agreed on inclusion/exclusion of studies 100% of the time. A decision to only double screen a proportion of titles and abstracts and papers was made based on the high level of agreement on screening suggesting that the inclusion and exclusion criteria were clear and that screening was accurate, the similar approach to double coding used in other reviews (Furuta, Sandall, & Bick, 2012; Lucas, Olander, Ayers, & Salmon, 2019; Sambrook Smith, Lawrence, Sadler, & Easter, 2019).

A data extraction sheet was designed and used to extract relevant information from the full texts. This included: (1) measure used; (2) language of the measure; (3) country; (4) number of participants; (5) participant group; (6) participant demographics; (7) design of the study; (8) norming data for the measure; (9) cut-off scale of the measure; (10) how the measure was administered. The

primary outcome measure was reduction in FOC. Other outcome measures were self-efficacy, obstetric outcomes and childbirth experience. Data extraction for all studies was completed by RW. BRG completed data extraction for a proportion of studies (10%, $n = 7$). Agreement was high (88%). RW and BRG extracted effect sizes and statistics for the meta-analysis in duplicate.

Quality assessment

The Cochrane Risk of Bias Tool (Higgins *et al.*, 2011) was used to assess quality. Due to the nature of psychological intervention studies, both participants and personnel were aware of the intervention; therefore, as all studies would score negatively on the performance bias items, this was not assessed. The tool was adapted so that each bias criterion could be answered as 'yes', 'no' or 'not applicable' (n/a). Items that were scored yes were assigned a score of 1, items that were scored no were assigned a score of 0. These were then averaged (excluding the answers scored n/a) and multiplied by 100. Studies that scored between 0 and 33 were labelled as having a high risk of bias, those scoring between 34 and 66 were labelled as having a medium risk of bias, studies that scored between 67 and 100 were labelled as having a low risk of bias.

Data analysis

Studies were synthesised narratively then meta-analysis used to determine whether FOC interventions were effective for two types of outcome: (1) FOC in late pregnancy; and (2) CS birth. Potential moderators of the effectiveness of FOC interventions were examined, i.e. risk of bias, country, study design, sample (only women with FOC *v.* all women; nulliparous *v.* mixed parity), intensity of the intervention and type of intervention. Studies were excluded from the meta-analysis if they did not report effect sizes for relevant outcomes ($n = 31$), were based on samples with men only ($n = 1$), did not have a control group ($n = 5$), or where the FOC intervention was provided intrapartum ($n = 1$). Effect sizes were calculated as Cohen's d using the difference between the pre- and post-test means to control for any pre-test group differences, and the posttest standard deviations (Lipsey & Wilson, 2001). For Haapio, Kaunonen, Arffman, and Åstedt-Kurki (2017), Cohen's d was derived from the odds ratio (Borenstein, Hedges, Higgins, & Rothstein, 2009). Meta-analysis was carried out using R where the computation of a Q statistic was made. To see if publication bias was of any concern in this analysis, a funnel plot and trim and fill analysis (Duval & Tweedie, 2000) were carried out using R.

Results

Study characteristics

Searches identified a total of 4474 citations. Hand searches of reference lists of key papers identified a further 19 papers. After removing duplicates and screening through abstracts, titles and full texts, 66 papers from 48 studies remained for inclusion in the review (Fig. 1). Participants within the studies varied. Only three studies reported ethnicity, all recruited white women (range: 17–71%) and two studies reported recruiting Black women (range: 25–50%). University education was reported by 19 studies, 12 of these reported more than half of participants having completed university. Of the 18 studies that reported marital status, between 57% and 100% of the participants were married

or cohabiting. Most papers ($n = 27$) used the Wijma Delivery Expectance Questionnaire (WDEQ-A) to measure FOC. Scores of ≤ 37 are indicative of mild fear, scores of 38–65 moderate fear, scores of 66–84 severe fear, and scores of ≥ 85 clinical fear (Wijma, Wijma, & Zar, 1998). Average WDEQ-A scores ranged from 29.7 to 130.

One paper described their outcome variable as 'tokophobia' (Pour-Edalati, Moghadam, Shahesmaeili, & Salehi-Nejad, 2019) and one paper used the terms FOC and tokophobia interchangeably (Bulez, Ceber Turfan, & Sogukpinar, 2019). The remaining studies used FOC as their outcome variable. The majority of papers examined FOC interventions with women (see online Supplementary Table S2). The exceptions were two studies with fathers (Bergström, Rudman, Waldenström, & Kieler, 2013; Ryding *et al.*, 2018) and one with couples (Ahmadi *et al.*, 2018). Studies were mostly conducted in non-English speaking countries: Iran ($n = 8$), Sweden ($n = 9$), Turkey ($n = 9$). Sample sizes ranged from 10 to 1887. Three studies had a high risk of bias, 30 had a medium risk of bias and 15 had a low risk of bias (online Supplementary Table S3).

Interventions identified

Interventions were grouped into six broad categories:

- Cognitive/cognitive behavioural (11 papers from six studies): these were interventions which used strategies to change cognitions (e.g. psychoeducation, thought restructuring, problem solving) and behaviours (e.g. exposure, relaxation).
- Other talking therapies (16 papers from 12 studies): interventions that used therapeutic conversation (e.g. counselling, haptotherapy, psychotherapy).
- Antenatal education (18 papers from 13 studies): these were interventions where the main focus was education about pregnancy and birth.
- Enhanced midwifery care (six papers from three studies) comprised continuity of carer or a midwife-led visit to delivery suite.
- Alternative interventions (13 papers from 12 studies): interventions involving specific therapies such as hypnobirthing, stand-alone relaxation, Pilates, art therapy.
- Interventions during labour (two papers from two studies) used intrapartum music or emotional freedom technique (EFT)

Cognitive/cognitive behavioural

Six studies examined cognitive behavioural therapy (CBT) delivered online ($n = 2$; Baylis, Ekdahl, Haines, & Rubertsson, 2019; Hildingsson & Rubertsson, 2019; Larsson *et al.*, 2017; Larsson, Hildingsson, Ternström, Rubertsson, & Karlström, 2019; Nieminen *et al.*, 2015; Nieminen, Andersson, Wijma, Ryding, & Wijma, 2016; Rondung *et al.*, 2018) or face-to-face ($n = 4$; Kordi, Bakhshi, Masoudi, & Esmaily, 2017; Saisto, Salmela-Aro, & Nurmi, 2001; Sydsjö *et al.*, 2015; Uçar & Golbasi, 2019). Studies either had no control group ($n = 1$; Nieminen *et al.*, 2015, 2016) or compared the intervention to other talking therapies ($n = 2$; Baylis *et al.*, 2019; Hildingsson & Rubertsson, 2019; Larsson *et al.*, 2017, 2019; Rondung *et al.*, 2018; Saisto *et al.*, 2001) or standard medical care (SMC; $n = 3$; Kordi *et al.*, 2017; Sydsjö *et al.*, 2015; Uçar & Golbasi, 2019). Two studies had a low risk of bias and the rest medium.

Three studies, including online CBT, found a reduction in FOC symptoms (Kordi *et al.*, 2017; Nieminen *et al.*, 2016; Uçar & Golbasi, 2019). However, there was no impact of CBT on

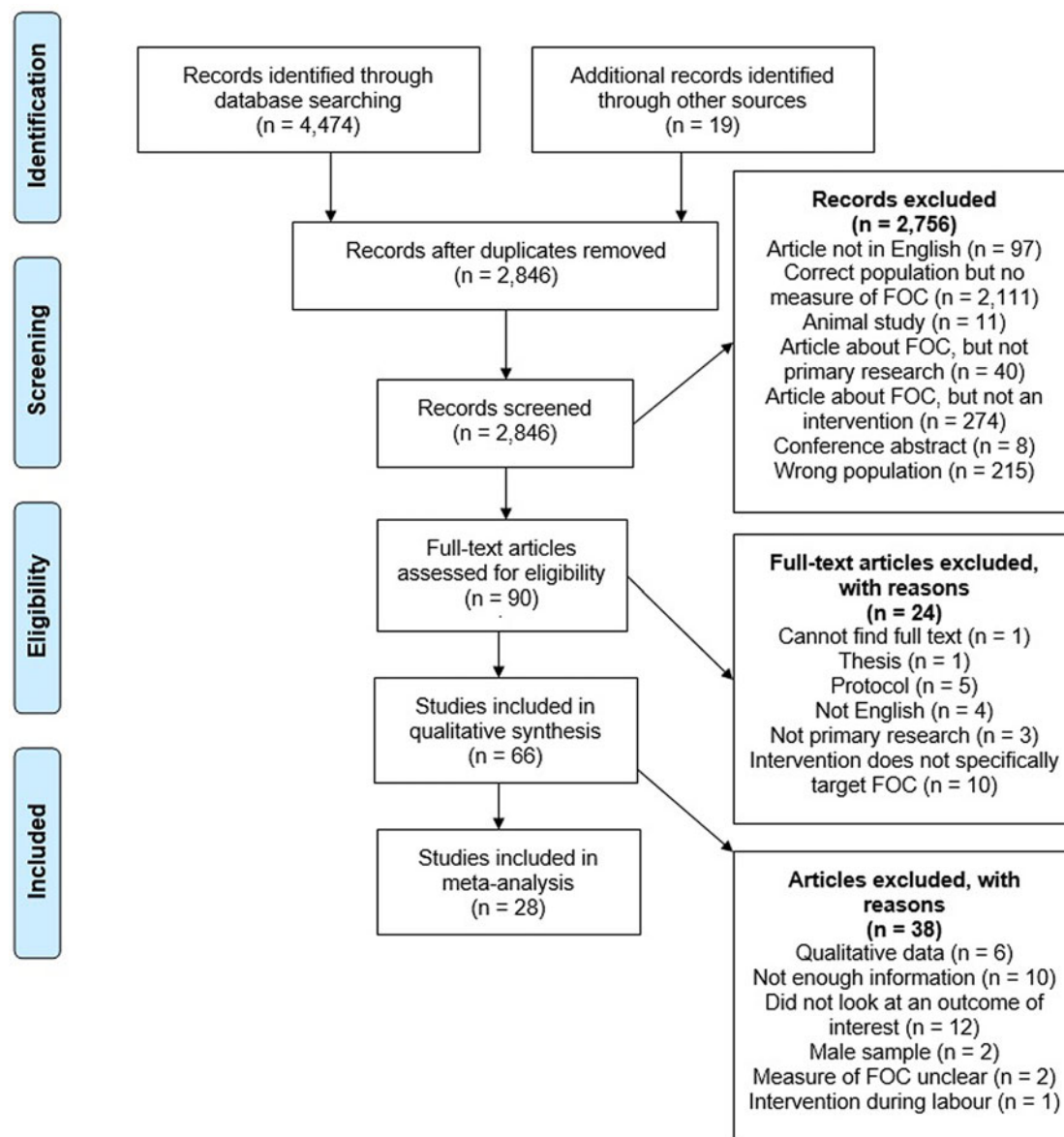


Fig. 1. PRISMA Flow Diagram.

birth preference, birth mode (Larsson et al., 2017) or birth experience (Hildingsson & Rubertsson, 2019), in fact one study found CBT was associated with an increase in negative birth outcomes (Sydsjö et al., 2015).

Other talking therapies

Twelve studies examined other talking therapies (excluding CBT) delivered face-to-face ($n = 11$; Ahmadi et al., 2018; Andaroon, Kordi, Kimiaei, & Esmaeily, 2017; Halvorsen, Nerum, Sørli, & Øian, 2010; Henriksen, Borgen, Risløkken, & Lukasse, 2018; Klabbers, Paarlberg, & Vingerhoets, 2018; Klabbers, Wijma, Paarlberg, Emons, & Vingerhoets, 2019; Larsson, Karlström, Rubertsson, & Hildingsson, 2015; Nerum, Halvorsen, Sørli, & Øian, 2006; Ryding, Persson, Onell, & Kvist, 2003; Sjogren, 1998; Soltani, Eskandari, Khodakarami, Parsa, & Roshanaei, 2017; Sydsjö et al., 2012) or over the telephone ($n = 1$; Fenwick et al., 2015; Toohill et al., 2014; Toohill, Callander, Gamble, Creedy, & Fenwick, 2017; Turkstra et al., 2017). These approaches

are a very diverse group, drawing on different theoretical frameworks including generic counselling, haptotherapy and psychotherapy. Studies either had no control group ($n = 2$) or compared other talking therapies to SMC ($n = 7$) or no intervention ($n = 3$). Two studies had a low risk of bias, one had high risk and the remaining medium risk.

Three studies, including other talking therapies carried out over the phone, found a reduction in FOC (Andaroon et al., 2017; Soltani et al., 2017; Toohill et al., 2014). However, two studies found no change in women's FOC after receiving other talking therapy (Larsson et al., 2015; Ryding et al., 2003). All three studies that looked at the impact of other talking therapies on birth preference found a reduction in desire for a CS (Fenwick et al., 2015; Halvorsen et al., 2010; Nerum et al., 2006). Three studies found other talking therapy was associated with a lower risk of CS (Ahmadi et al., 2018; Fenwick et al., 2015; Toohill et al., 2017); however, two studies found an increased risk of CS (Henriksen et al., 2018; Sydsjö et al., 2012).

Antenatal education

Thirteen studies examined antenatal education classes. All but two (Khedr & Eldeen, 2017; Kulkarni, Wright, & Kingdom, 2014) interventions were delivered face-to-face. Studies either had no control group ($n=2$; Khedr & Eldeen, 2017; Kulkarni et al., 2014) or compared antenatal education to SMC ($n=10$) or no intervention ($n=1$; Taheri, Mazaheri, Khorsandi, Hassanzadeh, & Amiri, 2014). Five studies were RCTs with low risk of bias (Bergström et al., 2013; Haapio et al., 2017; Masoumi et al., 2016; Ozdemir, Cilingir, Ilhan, Yildiz, & Ohanoglu, 2018; Rouhe et al., 2015).

All but one study (Masoumi et al., 2016) found that antenatal education was associated with a reduction of FOC in women (El-Malky, El-Homosy, Ashour, & Shehada, 2018; Gökçe İsbir, İnci, Önal, & Dikmen-Yıldız, 2016; Haapio et al., 2017; Karabulut, Coşkun Potur, Doğan Merih, Cebeci Mutlu, & Demirci, 2016; Kizilirmak & Başer, 2016; Serçekuş & Başkale, 2016; Taheri et al., 2014) and men (Bergström et al., 2013). Antenatal education was also associated with a change in birth preferences (Ozdemir et al., 2018), an increased likelihood of having a spontaneous vaginal birth (Rouhe et al., 2015) and a more positive birth experience (Rouhe et al., 2013).

Enhanced midwifery care

Three studies examined enhanced midwifery care, including a continuity of care model ($n=2$; Hildingsson, Rubertsson, Karlström, & Haines, 2018; Hildingsson, Karlström, Rubertsson, & Haines, 2019; Hildingsson et al., 2019; Lyberg & Severinsson, 2010a, 2010b) or a midwife-led visit to a delivery suite (Sydsjö et al., 2014). One study had a high risk of bias, and the remaining had a medium risk of bias. The results showed that continuity of care was evaluated positively by women (Lyberg & Severinsson, 2010a, 2010b) and was associated with reduced FOC, increased satisfaction with care (Hildingsson et al., 2018; Hildingsson et al., 2019a) and an improved birth experience (Hildingsson, Rubertsson, Karlström, & Haines, 2019b). The impact of visiting the delivery suite with a midwife prior to labour was associated with a shorter duration of labour for multiparous women. However, rate of emergency CS was higher in the intervention group (Sydsjö et al., 2014).

Alternative interventions

Twelve studies examined alternative interventions, including relaxation (e.g. guided relaxation, meditation/mindfulness, hypnobirthing; Baleghi, Akerdi, & Pasha, 2016; Bulez et al., 2019; Byrne, Hauck, Fisher, Bayes, & Schutze, 2014; Fisher et al., 2012; Hunter et al., 2011; Pour-Edalati et al., 2019), exercise ($n=2$; Guder, Yalvac, & Vural, 2018; Guskowska, 2014), art therapy ($n=2$; Sezen & Ünsalver, 2019; Wahlbeck, Kvist, & Landgren, 2018) or group psychodynamic (Saisto, Toivanen, Salmela-Aro, & Halmesmäki, 2006), role play (Navaee & Abedian, 2015) and heart rate monitoring (Narita, Shinohara, & Kodama, 2018). Ten studies had control groups; no studies were RCTs.

Relaxation-style interventions and art therapy were associated with a decrease in FOC (Baleghi et al., 2016; Bulez et al., 2019; Byrne et al., 2014; Fisher et al., 2012; Pour-Edalati et al., 2019; Sezen & Ünsalver, 2019; Wahlbeck et al., 2018); however, all but one of these studies had a medium or high risk of bias. Results for the remaining interventions were less clear. Role play reduced FOC; however, a reduction in FOC was also found in the control group (Navaee & Abedian, 2015). One exercise intervention including yoga, Pilates and body ball found no

impact on FOC (Guskowska, 2014) whereas another Pilates course was found to reduce FOC (Guder et al., 2018). Lastly, heart rate variability biofeedback was associated with lower FOC over time (from 32–34 to 36–67 weeks) (Narita et al., 2018).

Interventions during labour

Two interventions were carried out during labour, one had low risk of bias and the other had medium risk. One used an RCT to evaluate the effect of EFT [exposure therapy and somatic stimulation using acupressure points (i.e. tapping) compared to breathing awareness or SMC and found a reduction of FOC in the intervention group (Irmak Vural & Aslan, 2019)]. Another RCT using marching songs and cheerful music during labour found no impact on FOC, sense of power and self-control (Phumdoung, Youngvanichsate, & Wongmuneeworn, 2011).

Quantitative meta-analysis

Characteristics of studies

Twenty-eight papers from 22 studies were included in the meta-analysis (see online Supplementary Table S4). The majority of papers (79%) were carried out in Scandinavian (Sweden or Finland) or Middle-Eastern (Iran, Turkey or Egypt) countries. A variety of types of intervention were employed, the most frequent being antenatal education ($n=10$; 36%). The intensity of interventions varied from six or more sessions (high intensity) to two or fewer sessions. Control groups were mostly routine care, although in six papers the control was some form of counselling. Design of the studies included randomised controlled trials ($n=12$), quasi-experimental ($n=14$) and a pilot study ($n=2$). Papers were recent with a median publication year of 2017.

Publication bias

The trim and fill analysis and funnel plot showed that there were 0 estimated studies missing indicating that there is no significant funnel plot asymmetry (model estimate = -1.3673 ; s.e. = 0.3006 , $p < 0.0001$; $\tau^2 = 1.837$; s.e. = 0.600). As can be seen from Fig. 2, studies are evenly distributed around the central effect size and there is no evidence of studies that have non-significant or opposite results being omitted.

Types of outcome

For the majority of papers ($n=23$), the outcome was a measure of FOC. The remaining papers ($n=12$) recorded the mode of childbirth (whether or not a CS). There was overlap between papers (see online Supplementary Table S4).

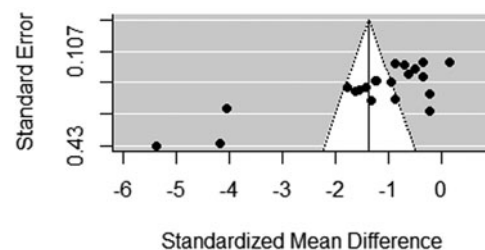


Fig. 2. Publication bias funnel plot.

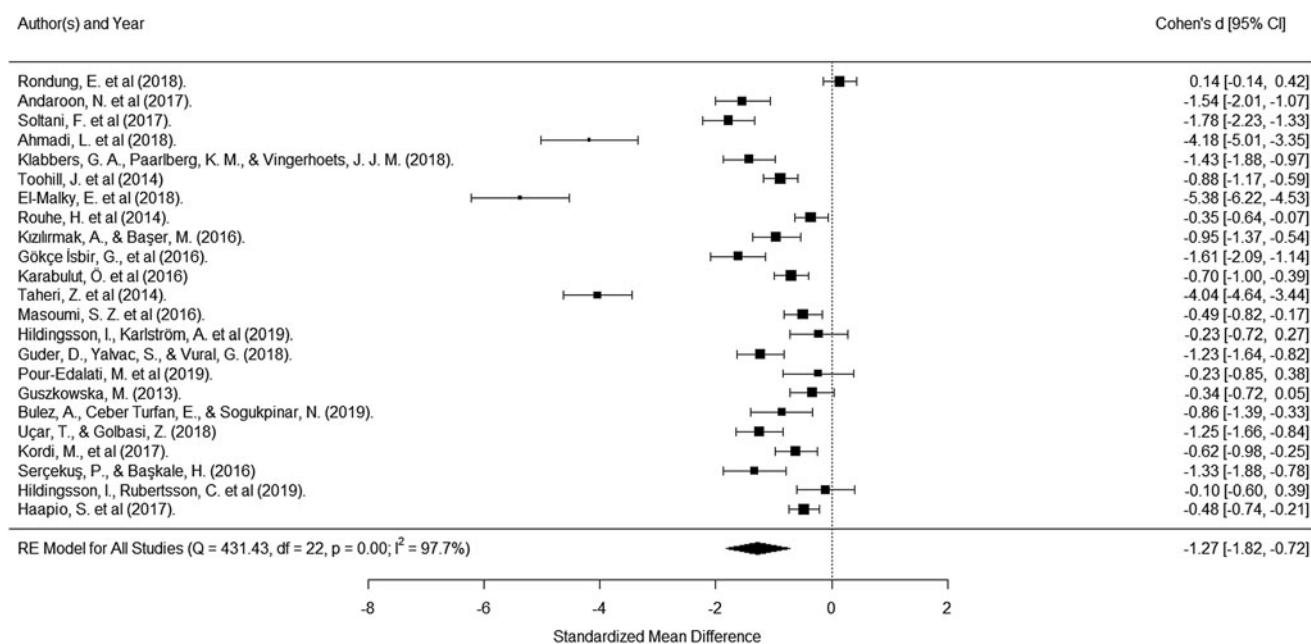


Fig. 3. Fear of childbirth forest plot.

Fear of childbirth

Most papers ($n=16$) used the Wijma Delivery Expectancy/Experience Questionnaire to measure FOC (Wijma et al., 1998) (W-DEQ).

The overall mean effect size was -1.27 ($z = -4.53$, $p < 0.0001$) indicating that women in the intervention group experienced much less FOC compared to those in the control group. There was, however, considerable heterogeneity in the size of the effect reported ($Q = 431.43$, $p < 0.0001$; $I^2 = 97.7$). All papers reported a lower FOC following intervention except Rondung et al. (2018) (Fig. 3 and Table 1). In three studies (Ahmadi et al., 2018; El-Malky et al., 2018; Taheri et al., 2014), the effect of the intervention was greater than four standard deviations, and an analysis of studentised residual indicated that these were significant outliers. When the analysis was re-run without these outliers, the overall effect was smaller but large and remained significant ($d = -0.80$, $z = -6.50$, $p < 0.0001$). Significant heterogeneity of effect sizes remained ($Q = 138.80$, $p < 0.0001$; $I^2 = 87.3$).

Examination of moderators found country was the only moderator of effectiveness (Table 2). Studies in Middle-Eastern countries reported much larger effect sizes ($d = -1.54$, $z = -2.23$, $p = 0.026$, $n = 14$) than those from Scandinavian countries ($d = -0.20$, $z = -0.34$, $p = 0.731$, $n = 5$). Whether the intervention was antenatal education or other talking therapies did not significantly moderate the effect. It was not possible to examine other intervention types because there were too few studies.

Mode of delivery

Twelve studies recorded whether birth was by CS (Fig. 4 and Table 3). Overall, the odds of a CS were lower in the intervention group compared to the control group, but this difference was not statistically significant (OR = 0.80, $z = -1.85$, $p = 0.065$). The test for homogeneity was also not significant [Q ($df = 11$) = 15.23, $p = 0.172$]. Examination of studentised residuals indicated there were no significant outliers.

Two significant modifiers were found: country and type of intervention (Table 4). In Scandinavian countries, the odds of a CS were significantly lower in the intervention group (OR 0.66, 95% CI 0.49–0.90), whereas in Middle-Eastern countries, women in the intervention group were more likely to have a CS (OR 1.07, 95% CI 0.76–1.52). Those who undertook talking therapies were less likely to have a CS (OR 0.48, 95% CI 0.48–0.90) compared to those who received antenatal education (OR 1.01, 95% CI 0.78–1.31).

Discussion

Main findings

This review aimed to identify interventions that reduced FOC and CS rates. The review identified 66 papers from 48 studies that investigated six types of intervention. Most studies were carried out in non-English speaking countries and only two recruited fathers-to-be. The quality of the studies varied with 30 out of 48 having a medium risk of bias.

Results from the meta-analysis suggest that most interventions, regardless of the approach, reduce FOC. This is in line with results from the qualitative synthesis which found that within each intervention approach, the majority of studies found a reduction in FOC. The studies that did not find a reduction of FOC were counselling run by midwives (Ryding et al., 2003), self-reported counselling (Larsson et al., 2015), antenatal education classes run by midwives with a focus on physical wellbeing (Masoumi et al., 2016), an exercise intervention including yoga, pilates and body ball (Guszkowska, 2014) and listening to marching songs during labour (Phumdoung et al., 2011).

The effect of interventions on CS rates was varied and insignificant overall, with studies of CBT (Larsson et al., 2019) finding no reduction in CS rates, and enhanced midwifery care finding increased likelihood of CS (Sydsjö et al., 2014). Talking therapies were more consistently associated with reduced CS rates whether

Table 1. Effect sizes for fear of childbirth interventions on FOC reduction

Author and year	<i>n</i>	Country	Risk of bias	Effect size Cohen's <i>d</i>	95% CI
CBT					
Kordi et al. (2017)	120	Iran	Low	-0.62	-0.98 to -0.25
Rondung et al. (2018)	258	Sweden	Low	0.14	-0.14 to 0.42
Uçar and Golbasi (2019)	111	Turkey	Medium	-1.25	-1.66 to -0.84
			Range:	0.14, -0.62	
Other talking therapies					
Ahmadi et al. (2018)	71	Iran	Medium	-4.18	-5.01 to -3.35
Andaroon et al. (2017)	93	Iran	Low	-1.54	-2.01 to -1.07
Klabbers et al. (2018)	134	The Netherlands	Medium	-1.43	-1.88 to -0.97
Soltani et al. (2017)	106	Iran	Medium	-1.78	-2.23 to -1.33
Toohill et al. (2014)	339	Australia	Low	-0.88	-1.17 to -0.59
			Range:	-0.88, -4.18	
Antenatal education					
El-Malky et al. (2018)	100	Egypt	High	-5.38	-6.22 to -4.53
Haapio et al. (2017)	659	Finland	Low	-0.48	-0.74 to -0.21
Gökçe İsbir et al. (2016)	90	Turkey	Medium	-1.61	-2.09 to -1.14
Karabulut et al. (2016)	192	Turkey	Medium	-0.70	-1.00 to -0.39
Kizilirmak and Başer (2016)	99	Turkey	Medium	-0.95	-1.37 to -0.54
Masoumi et al. (2016)	160	Iran	Low	-0.49	-0.82 to -0.17
Rouhe et al. (2015)	330	Australia	Low	-0.35	-0.64 to -0.07
Serçekuş and Başkale (2016)	63	Turkey	Medium	-1.33	-1.88 to -0.78
Taheri et al. (2014)	130	Iran	Low	-4.04	-4.64 to -3.44
			Range:	-0.35, -5.38	
Enhanced midwifery care					
Hildingsson et al. (2019a) ^a	70	Sweden	Medium	-0.23	-0.72 to 0.27
Hildingsson and Rubertsson (2019) ^a	70	Sweden	Medium	-0.10	-0.60 to 0.39
			Range:	-0.10, -0.23	
Alternative interventions					
Bulez et al. (2019)	60	Turkey	High	-0.86	-1.39 to -0.33
Guder et al. (2018)	108	Cyprus	Medium	-1.23	-1.64 to -0.82
Guszkowska (2014)	109	Poland	Medium	-0.34	-0.72 to 0.05
Pour-Edalati et al. (2019)	41	Iran	Medium	-0.23	-0.85 to 0.38
			Range:	-0.23, -0.86	

^aResults from the same study.

delivered by telephone (Fenwick et al., 2015; Toohill et al., 2017) or face-to-face (Ahmadi et al., 2018). Results from the moderator analysis support this, in that women who undertook talking therapies were less likely to have a CS compared to those who received antenatal education. This suggests talking therapies may be potentially effective for reducing CS rates in women with FOC. However, the majority of these studies had a medium risk of bias and the results were not completely consistent with two studies finding that other talking therapies were associated with an increased risk of CS (Henriksen et al., 2018; Sydsjö et al., 2012). More research is therefore needed.

Interpretation

The results from the qualitative and quantitative analysis of this review suggest that most intervention approaches investigated reduce FOC. Only five studies across all intervention groups found null results in terms of FOC reduction, compared to 25 studies that found a reduction in FOC. It may therefore be the delivery of an intervention, rather than the theoretical underpinning that makes it effective at reducing FOC. This is supported by a recent meta-synthesis of women's experiences of interventions for FOC (O'Connell, Khashan, & Leahy-Warren, 2020) that found interventions with a woman-centred ethos, where women

Table 2. Moderators of effect of intervention on fear of childbirth ($k = 23$ unless otherwise stated)

Moderators	QM	QE
Risk of bias	0.18	413.17**
Country ($k = 19$)	4.98*	303.00**
Whether or not RCT	1.10	302.41**
Sample: only women with FOC v. all women	2.37	407.84**
Sample: nulliparous v. mixed parity ($k = 21$)	1.43	408.31**
Intensity of intervention ($k = 19$)	0.63	358.19**
Other talking therapies v. antenatal education ($k = 14$)	0.09	314.46**

* $p < 0.05$; ** $p < 0.01$.

felt listened to and where a trusting relationship was able to develop were crucial for women to move from fear to ownership of childbirth. Further, most women felt empowered when they attended FOC interventions, and this was facilitated by supportive alliances, education and birth choices. It could therefore be suggested that providing women with a supportive space to explore their fear is more important than providing a specific approach in terms of reducing FOC. However, more research is needed to understand whether this is the case.

Another potential reason for this finding is that there is a wide range of different fears involved in FOC as well as individualised responses (Wigert et al., 2020). Further, studies do not always differentiate between primary tokophobia (women who have not given birth before) and secondary (women who have given birth before) tokophobia. This is an important distinction to make, because secondary tokophobia usually develops after a previous traumatic birth (Bhatia & Jhanjee, 2012) and, although not specific to childbirth, there is an evidence base about how trauma and PTSD can be treated [National Institute for Health and Care Excellence (NICE), 2018].

The findings from previous systematic reviews that identified interventions with an educational component as being promising at reducing FOC (Moghaddam Hosseini et al., 2018; Stoll et al., 2018; Striebich et al., 2018) warrants further exploration. This review found that interventions with an educational component were the most studied ($n = 19$ studies), but not the most effective. It is therefore possible that the findings from previous systematic reviews can be explained by the fact that interventions with an educational component appeared to be most promising because they had been studied more. However, it is not clear if this is the only explanation, therefore more high-quality RCTs are needed to examine the effectiveness of different types and components of interventions on FOC.

The varied and insignificant effect of FOC interventions on CS rates is unsurprising given the many physiological, psychosocial, contextual, organisational and cultural factors that influence whether a woman wants, requires or has a CS. This is illustrated by the finding that country of study moderated the effect of FOC interventions on CS rates, suggesting cultural and organisational context are important. For example, FOC interventions were found to reduce CS rates amongst women in Scandinavian countries and increase rates in Middle Eastern countries (Iran, Egypt and Turkey). This is perhaps reflective of maternity care in these countries. Iran and Egypt both provide a medicalised model of maternity care, with high mortality and very little antenatal education (Aghlmand et al., 2008; Choices and Challenges in Changing Childbirth Research Network, 2005; El-Kurdy, Hassan, Hassan, & El-Nemer, 2017; TorkZahrani, 2008). Further, the rates of CS are higher in these countries at 48–52% (Azami-Aghdash, Ghojzadeh, Dehdilani, Mohammadi, & Asl Amin Abad, 2014; Ministry of Health and Population et al., 2015; Santas & Santhas, 2018) than in Scandinavian countries (15–20%) (Pyykönen et al., 2017). Additionally, it is important to note that in some cases, a planned CS can improve women’s birth experience, such as where there is not sufficient time for FOC intervention, or where FOC has not improved following intervention. A planned CS may be the next most appropriate

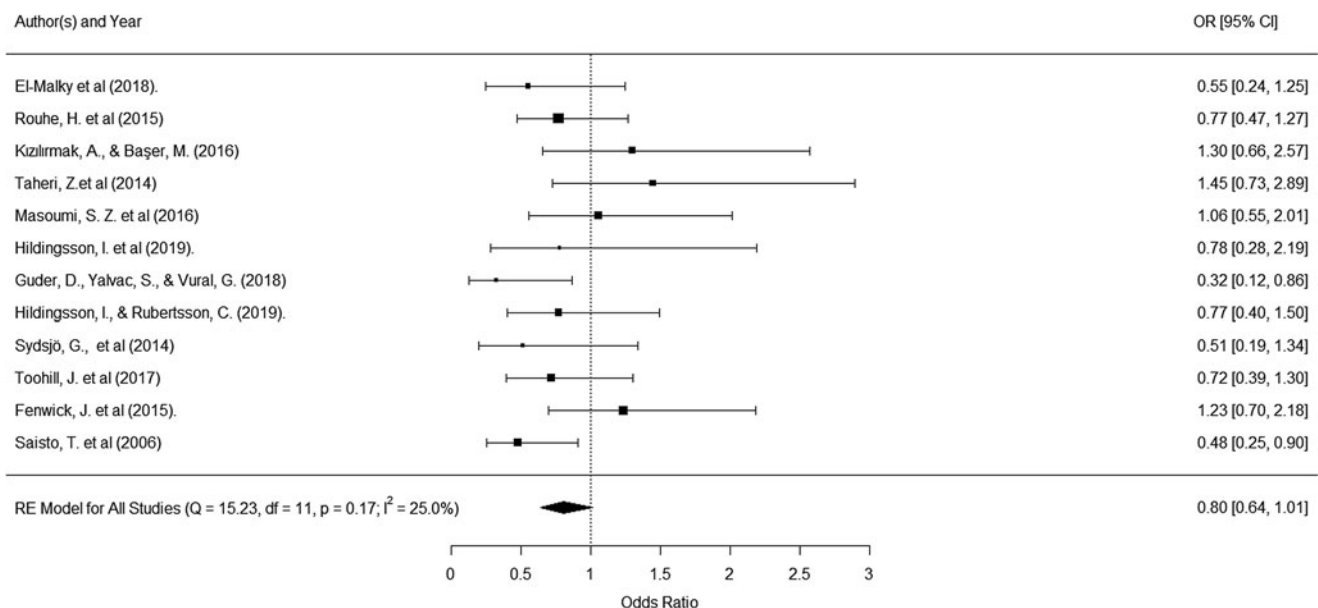


Fig. 4. Caesarean section by choice forest plot.

Table 3. Effect sizes for fear of childbirth interventions on CS rate reduction

Author and year	<i>n</i>	Country	Risk of bias	Effect size (Cohen's <i>d</i>)	95% CI
CBT					
Hildingsson and Rubertsson (2019)	258	Sweden	Low	0.77	0.40–1.50
Sydsjö <i>et al.</i> (2015)	612	Sweden	Medium	0.51	0.19–1.34
Range:				0.51, 0.77	
Other talking therapies					
Fenwick <i>et al.</i> (2015) ^a	184	Australia	Low	1.23	0.70–2.18
Toohill <i>et al.</i> (2017) ^a	184	Australia	Low	0.72	0.39–1.30
Range:				0.72, 1.23	
Antenatal education					
El-Malky <i>et al.</i> (2018)	100	Egypt	High	0.55	0.24–1.25
Kizilirmak and Başer (2016)	99	Turkey	Medium	1.30	0.66–2.57
Masoumi <i>et al.</i> (2016)	160	Iran	Low	1.06	0.55–2.01
Rouhe <i>et al.</i> (2015)	330	Finland	Low	0.77	0.47–1.27
Taheri <i>et al.</i> (2014)	130	Iran	Low	1.45	0.73–2.89
Range:				0.55, 1.45	
Enhanced midwifery care					
Hildingsson <i>et al.</i> (2019a)	70	Sweden	Medium	0.78	0.28–2.19
Range:				0.78	
Alternative interventions					
Guder <i>et al.</i> (2018)	108	Cyprus	Medium	0.32	0.12–0.86
Saisto <i>et al.</i> (2006)	187	Finland	Medium	0.48	0.25–0.90
Range:				0.32, 0.48	

^aResults from the same study.

Table 4. Moderators of effect of intervention on fear of childbirth (*k* = 12 unless otherwise stated)

Moderators	QM	QE
Risk of bias	1.61	13.34
Country (<i>k</i> = 9)	4.11*	5.53
Whether or not RCT (<i>k</i> = 11)	2.36	9.77
Sample: only FOC <i>v.</i> all women	0.71	14.14
Sample: nulliparous <i>v.</i> mixed parity (<i>k</i> = 11)	1.44	12.62
Intensity of intervention (<i>k</i> = 8)	1.65	11.19
Other talking therapies <i>v.</i> antenatal education (<i>k</i> = 7)	4.52	5.31

p* < 0.05; *p* < 0.01.

clinical step and can allow for women to feel a sense of control of their birth experience. This suggests individual, cultural and organisational norms may influence whether FOC interventions affect CS rates, and the wider care environment should be taken into consideration when selecting a FOC intervention.

Strengths and limitations

A strength of this review is the broad inclusion criteria, meaning all identified studies on FOC interventions were included.

Another strength is the meta-analysis which provides a novel quantitative understanding of the effectiveness of FOC interventions in more robustly designed studies. Limitations of the review include the decision to only double screen 10% of abstracts and full texts. This may have meant some papers were missed; however, the high concordance of the double screening makes this seem unlikely. Further limitations are the low number of studies with different intervention types that could be included in the meta-analysis. There was great heterogeneity of the effect sizes for the FOC outcome across the studies (e.g. *Q* = 431.43; *Q* = 138.80). This makes interpretation of the results more difficult. Classification of interventions was challenging because they were complex interventions with multiple components. Some components could overlap between intervention categories. Components of interventions were also minimally described in many papers. Similarly, the methodological quality of studies was varied, with 38 out of the 48 studies had medium to high risk of bias. The way studies were carried out was also variable. For example, different outcome measures were used making it difficult to compare the baseline levels of FOC in all studies. Furthermore, those that used the WDEQ-A and were comparable recruited women with a range of WDEQ-A scores making results difficult to interpret (*M* = 29.7–130). Additionally, where participant demographics were reported, women were often highly educated, married and white, meaning these results may not be generalisable to those from more marginalised populations. Due

to these limitations, the results from these studies should be interpreted cautiously.

Conclusion

Overall, this review suggests that interventions for FOC are effective in reducing FOC but have variable effects on CS rates. FOC interventions do not affect CS rates overall, but this is influenced by cultural and organisational context. High-quality RCTs of different FOC interventions that not only evaluate type of intervention but also determine which components of interventions are most effective for particular presentations are needed in order to design optimal FOC interventions.

Supplementary material. The supplementary material for this article can be found at <https://doi.org/10.1017/S0033291721002324>

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